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The utility of fistulography in the diagnosis of thyroglossal duct cyst with fistulous tract: Case report and literature review

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Thyroglossal duct cyst (TGDC) is one of the most common congenital, midline, cervical lesions originating from an embryonic thyroglossal duct remnant. It is usually diagnosed clinically. Imaging is used to confirm the clinical diagnosis, and fistulography is very helpful in the diagnosis and surgical planning for thyroglossal fistulous tract. Fistulographs show the direction, length, and anatomy of the remnant tracts and the small tracts connecting a TGDC to the tongue base at the foramen cecum. This report presents a case of TGDC in a 12-year-old boy and demonstrates the utility of fistulography imaging in the diagnosis of TGDC with fistulous tract. In this case, fistulography was performed after cannulating the external opening of the fistula. The patient underwent a resection, including thyroglossal tract removal and further excision of the mid portion of the hyoid bone (the Sistrunk procedure). The postoperative pathology report indicated thyroglossal duct remnants with no evidence of malignancy.

Introduction

TGDC is one of the most common congenital midline abnormalities of the neck, occurring in approximately 7% of the population and representing approximately 75% of congenital neck diseases (1). It results from the anomalous development and the involution failure in obliterating the embryogenic duct, produced during thyroid migration from the fourth through eighth weeks of gestation (2). The location of a TGDC may extend from the foramen cecum of the tongue base to the central portion of the hyoid bone. Most TGDCs occur between the thyroid gland and the hyoid bone, and the most common complications of TGDC are infection and fistula formation. There may be small tracts and branches connecting the external opening of the fistula on the skin to the foramen cecum of the tongue base.

The diagnosis of a TGDC is usually made by a clinical history and thorough physical examination, combined with imaging findings and the midline location. Patients will likely have an ultrasound to evaluate the mass further. Other imaging modalities, including magnetic resonance imaging (MRI) and computed tomography (CT) play a supplementary role in the diagnosis of TGDC in children, due to the disadvantages of general anesthesia and ionizing radiation. Fistulography may show the course of the tract, and it is useful for the patient with a history of recurrent lateral neck abscess when an internal sinus opening is suspected. However, this technique is seldom used as a routine basis for the diagnosis of thyroglossal fistula and has seldom been reported in children.

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Case report

A 12-year-old boy was referred to the Department of Oral Surgery at Jilin University Stomatological Hospital with a recurrent mass and orificium fistulae in the anterior region of the neck. The patient presented with a history of a frequent painful swelling and a gradually enlarging midline neck mass over three months. At a local hospital, a surgical incision was performed, and antibiotics were used for the treatment. This resulted in sinus tract formation and an external fistulous opening, with drainage present at the skin of the neck.

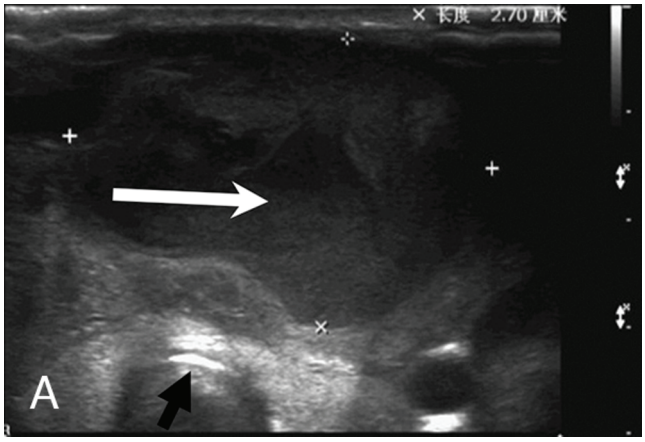

Fig. 1. Longitudinal grayscale ultrasound image in midline of the neck demonstrates a well-defined, oval-shaped, unilocular, anechoic lesion with thick wall (white arrow) located just above the hyoid bone (black arrow). The posterior enhancement as a clue to the cystic nature of the lesion is indicated.

Clinical examination of the patient revealed a 40 × 40-mm neck mass that was painless, smooth, and well demarcated. The mass was located in the front of the midline neck and included a small discharging fistulous opening in the skin that drained fluid. It was mobile in the craniocaudal direction during swallowing and moved with protrusion of the tongue. The patient had no dyspnea, dysphagia, or dyspepsia.

Ultrasonography showed a normal-appearing thyroid gland and a midline, oval, unilocular, hypoechoic, debris-containing cystic mass. The mass, suspicious for an infected TGDC, had a clear boundary, a smooth wall, and posterior enhancement above the hyoid bone (Fig. 1). Fistulography demonstrated the location of the cyst and fistulous tracts anterior to the pharynx/hypopharynx and the cyst's relationship to the hyoid bone on the lateral and posterior-anterior radiographs. More specifically, the small sinus tracts ascending towards the tongue base were clearly identified, which allowed complete surgical resection and led to a definitive cure for the patient. The patient complained of a bitter taste after the injection of the contrast medium (76% compound meglumine diatrizoate), which confirmed

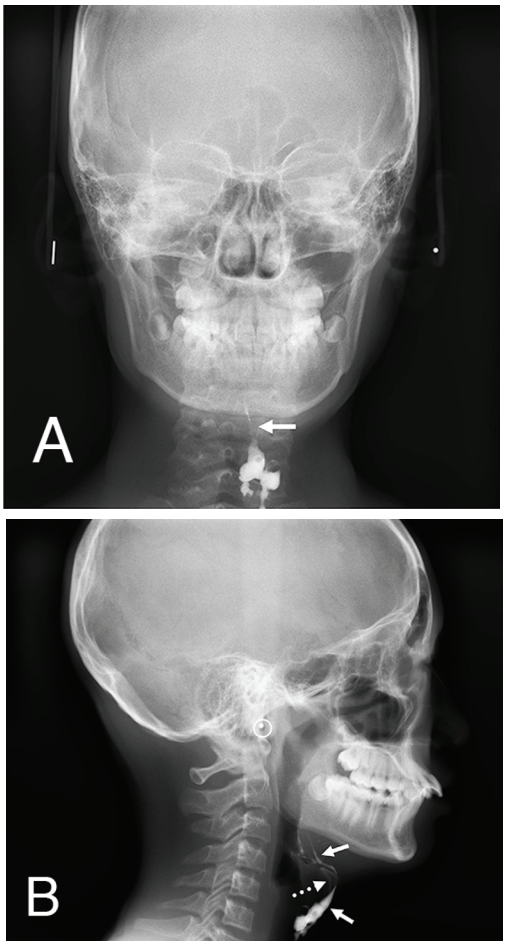

Fig. 2. Fistulograms. A. Posterior-anterior radiograph after injection of 76% compound meglumine diatrizoate into the duct through the fistulous opening of the skin in the midline of the neck. The thyroglossal tract can be identified, and the extent and anatomy of the remnant tracts were demonstrated. B. Lateral radiograph showing the location of the cyst, the small fistulous tracts ascending towards the tongue base, and their relationship to the hyoid bone. The white arrows show the tract of thyroglossal fistula, and the dotted white arrow shows the hyoid bone.

the fact that the fistulous tracts were communicating with the oral cavity (Fig. 2). CT and MRI were not performed as routine diagnostic imaging techniques, due to radiation and cost considerations.

The patient underwent a surgical excision treatment that involved complete removal of the cyst, the entire thyroglossal tracts, and the midpart of the hyoid bone (Fig. 3). A horizontal incision was made over the skin of the neck, and the tract was extended upward to the base of the tongue (the Sistrunk procedure). A specimen was sent for routine histopathological examination. The pathologic report described a cystic lesion with a fibrous wall lined by flattened epithelium, focal aggregates of chronic inflammatory cells, and thyroglossal duct remnants with no evidence of malig-

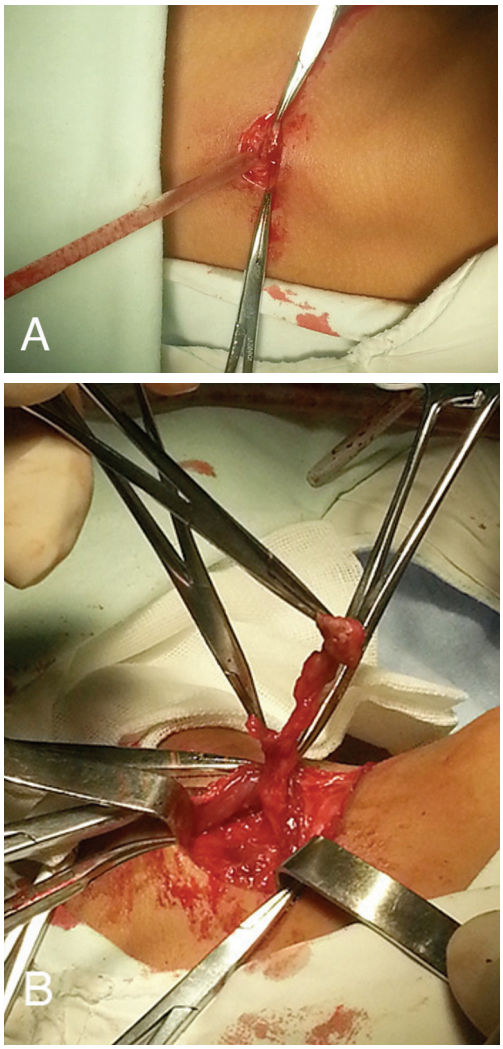

Fig. 3. At surgery. A. Surgical skin incision. B. Intraoperative view showing the cyst and thyroglossal fistulous tract.

nancy, which confirmed the provisional diagnosis of infected thyroglossal cyst (Fig. 4). The postoperative course was uneventful, and the patient had no recurrence at 2 months.

Discussion

TGDCs account for up to 70% of congenital abnormalities that occur in the anterior midline of the neck (3). They usually occur in the midline (75%) or slightly off midline (25%) of the anterior neck (4). They move upward during swallowing or on protrusion of the tongue due to their attachment to the tongue via the tract of thyroid descent. The exact frequency of the cyst is uncertain, but its incidence appears to be similar in both genders and is identified to be higher in children than in adults (5).

A TGDC can be easily diagnosed clinically when it presents with typical findings such as a small, round or oval lump in the center of the front part of the neck. Health history and physical examination are adequate to make a correct preoperative diagnosis. Imaging is used to confirm the clinical diagnosis, determine the extent of the lesion and its relationship to surrounding structures, and identify the presence of the fistulous tracts. Ultrasonography is the most frequent and preferred imaging modality for preoperative evaluation in suspected TGDC in children due to its comparable accuracy, low cost, noninvasive nature, and absence of ionizing radiation comparable to X-ray (6). However, ultrasound cannot reliably assess the infrahyoid TGDCs and cannot depict the relationship of the thyroglossal duct to the hyoid bone. Other imaging modali-

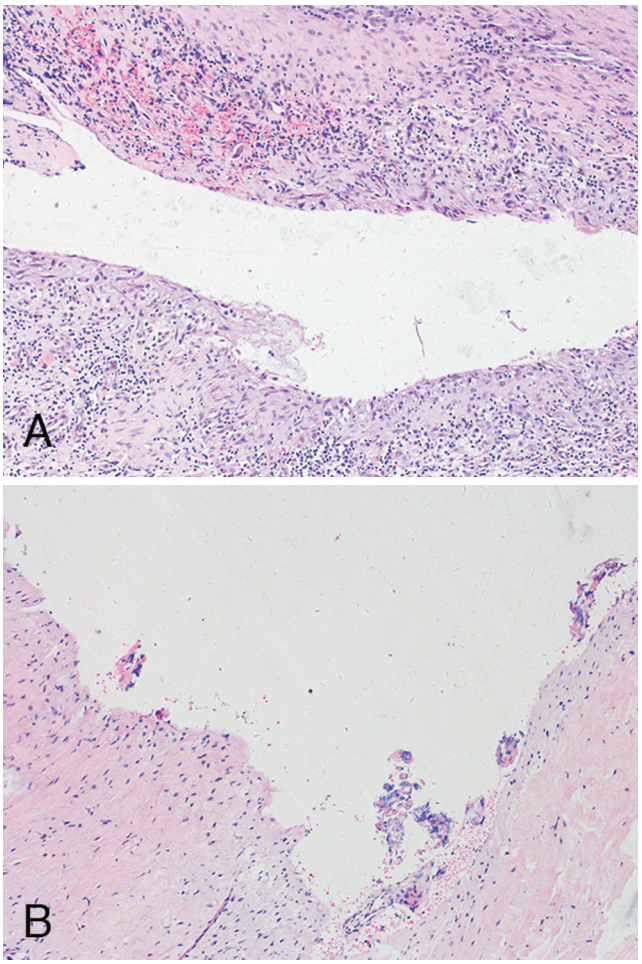

Fig. 4. The histopathological examination demonstrating a cystic lesion with a fibrous wall lined by flattened epithelium, focal aggregates of chronic inflammatory cells, and thyroglossal duct remnants with no evidence of malignancy. A. Chronic inflammatory cells and fibrocollagenous wall lined by epithelial cells of the tract (Hematoxylin and eosin, 100X original magnification). B. The lumen and wall of the cyst (Hematoxylin and eosin, 100X original magnification).

ties have been employed for the diagnosis of TGDCs. CT imaging of adult patients before treatment plays a supplementary role in more accurately depicting the anatomy of the lesion, particularly with large cysts (7, 8). MRI is the preferred modality for lesions at or close to the tongue base. In patients with cutaneous openings, fistulography may be performed. Fistulography is valuable and helpful in demonstrating the extent and anatomy of the residual fistulous tract. Ranga et al report a thyroglossal fistula fistulography using CT (9).

In the present case, ultrasound found a unilocular cystic lesion that had fluid and multiple internal echoes, which indicated an infected TGDC. Fistulography showed the location of the cyst and fistulous tracts and their relationship to the hyoid bone on the lateral and posterior-anterior radiographs. The residual suprahyoid tract up to the foramen cecum with side branches and the close association of the cyst and the hyoid bone were clearly visualized, which enabled surgeons to remove the entire thyroglossal tract remnant and reduce the possibility of postoperative recurrence.

In conclusion, in the preoperative imaging of a TGDC with fistulous tract, fistulography serves as an accurate, simple, fast, minimally invasive, and particularly useful diagnostic method for surgical planning. The identification of the thyroglossal tract before surgery using fistulography enables accurate local excision of the remaining fistulous tracts, ensures removal of the entire thyroglossal tract remnant, and reduces recurrence.

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